Clipping of ruptured aneurysm of lateral spinal artery associated with anastomosis to distal posterior inferior cerebellar artery: a case report

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DOI: https://doi.org/10.1016/j.wneu.2018.06.085

Elsevier Editorial System(tm) for World Neurosurgery

Manuscript Draft

Manuscript Number: WNS-18-2094

Title: Clipping of ruptured aneurysm of lateral spinal artery associated with anastomosis to distal posterior inferior cerebellar artery: a case report

Article Type: Case Report

Keywords: Cerebral angiography, Intracranial aneurysm, Spinal vascular malformation, Subarachnoid hemorrhage

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COVER LETTER

Manuscript title: Clipping of ruptured aneurysm of lateral spinal artery associated with anastomosis to distal posterior inferior cerebellar artery: a case report

Dear prof. Benzel,

Enclosed is our manuscript 'Clipping of ruptured aneurysm of lateral spinal artery associated with anastomosis to distal posterior inferior cerebellar artery: a case report' by M.R. Germans et al., which we would like to submit as a case report in World Neurosurgery.

The first author (M.R. Germans) has full access to the data and takes responsibility for the authenticity of the data. I, Menno R. Germans, certify that this manuscript is a unique submission and is not being considered for publication, in part or in full, with any other source in any medium.

We look forward to hearing from you at your earliest convenience.

With kind regards,

Menno R. Germans

On behalf of all authors

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We confirm that the manuscript has been read and approved by all named authors and that there are no other persons who satisfied the criteria for authorship but are not listed. We further confirm that the order of authors listed in the manuscript has been approved by all of us.

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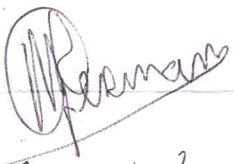
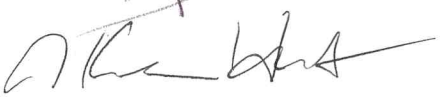
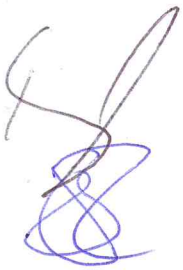
Menno R. Germans, MD, PhD		16/5/2018
Zsolt Kulcsar, MD		17/5/2018
Luca Regli, MD		17/5/2018
Oliver Bozinov, MD		17.5.18

Abbreviation list:

AICA	anterior inferior cerebellar artery
CT	computed tomography
DSA	digital subtraction angiography
ICG	indocyanine green
LSA	lateral spinal artery
PICA	posterior inferior cerebellar artery
SAH	subarachnoid hemorrhage
SCA	superior cerebellar artery

Highlights

- Aneurysms of the lateral spinal artery are rare and can cause subarachnoid hemorrhage
- Occlusion of a neighboring artery of the lateral spinal artery can activate the anastomotic network of the lateral spinal artery rapidly
- Follow-up of patients with an activated anastomotic network is warranted because of potential new aneurysm formation

Abstract

Arteries that anastomose with the posterior inferior cerebellar artery (PICA) can harbor aneurysms. We present a case of a patient who suffered a subarachnoid hemorrhage as a result from an aneurysm on the left lateral spinal artery (LSA) which anastomosed to the PICA. The aneurysm was surgically treated and the flow between the LSA and PICA was disrupted. The activated anastomotic network created a new anastomosis between the LSA and PICA which was seen at six months follow-up. Careful follow-up is warranted in patients who have an activated anastomotic network because they can potentially develop aneurysms on newly created anastomoses.

Background

A ruptured aneurysm of the distal posterior inferior cerebellar artery (PICA) accounts for only 0.5-3% of all subarachnoid hemorrhages (SAH). In comparison to other intracranial arteries, the PICA has a high rate of anatomical variations, making treatment of aneurysms on this artery challenging. Case reports have described an aneurysm on an artery that is anastomosed to the PICA. We report a patient with SAH, exclusively located in the cerebellomedullary cistern and a small cerebellar infarct. Preoperative angiography and surgical exploration revealed an aneurysm of the lateral spinal artery (LSA) with anastomosis to the distal PICA and occlusion of the proximal PICA.

Case description

History and examination

A 49-year old male patient was hospitalized with severe headaches, neck pain, dizziness, nausea and vomiting for approximately four days. His medical history included a deep venous thrombosis four years earlier and chronic venous insufficiency in both legs. General and neurological examination showed an obese patient without focal neurological deficits. Brain computed tomography (CT)

showed a contained subarachnoid hemorrhage in the left cerebellomedullary cistern with evidence for a small aneurysm-like vascular structure associated to the hematoma, without clear anatomical definition of the parent artery. Additionally, a small old cerebellar infarct was identified in the left PICA territory (**Figure 1**). The digital subtraction angiography (DSA) revealed a tortuous artery originating from the proximal V3 segment of the left vertebral artery, and running cranially along the dorsolateral medullary surface and showing a 3 mm aneurysm at the level of the tonsil (**Figure 2**). Distal from the aneurysm, the artery anastomosed with the left PICA. The artery was identified as the dilated lateral spinal artery (LSA), anastomosing with the PICA at its telovelotonsillary segment and filling further its vermian and hemispheric branches. The original, proximal segment of the PICA was not visualized on angiography. The distal hemispheric branches of the PICA were filled in a retrograde fashion through the anastomotic connections from left anterior inferior cerebellar artery (AICA) and superior cerebellar artery (SCA). Based on the cerebellar infarct and the lack of visualization of the PICA we hypothesized an earlier, dissection related occlusion of the PICA. Endovascular treatment was considered to have elevated risks due to the tortuous course and small diameter of the LSA with potential of perforation or permanent occlusion. On interdisciplinary discussion surgical exploration was decided.

Operation

The patient was positioned in left lateral modified park-bench position, as prone position was not favorable due to his obesity. A straight incision in the midline was planned. Suboccipital craniotomy with partial removal of the C1 arch was performed and the dura was opened in the midline. With careful intradural dissection the tonsillomedullary region was explored. The aneurysm on the LSA was identified. The inspection demonstrated an old occlusion of the proximal PICA up to the anastomotic level with the LSA, with a reperfused telovelotonsillary segment through this arterio-arterial connection **(Figure 3a)**. Indocyanine green (ICG) videoangiography showed a caudocranial flow from the LSA into the telovelotonsillary segment of the PICA (**Figure 3b and 3c**). The aneurysm

was secured by inflow obstruction with a single straight clip. Afterwards, the ICG videoangiography showed flow in the anastomosis via several pial medullary arteries and inverted (i.e. retrograde) flow into the telovelotonsillary segment of the PICA (**Figure 4**).

Postoperative course

The early postoperative course was uneventful and MR imaging showed no new ischemic deficits. Six months post-hemorrhage he was back to work without any complaints nor neurological deficits. A DSA at six months after surgery showed the complete occlusion of the aneurysm. The LSA showed a new anastomotic connection to the telovelotonsillary segment of the PICA with better anterograde flow in the hemispheric branches (**Figure 5**).

Conclusions

This case describes the activation of the pial medullary arterial anastomotic network after occlusion of the PICA at its origin, with the risk of developing an aneurysm and rupture, probably related to the increased flow related hemodynamic changes in these originally tiny vessels. The aneurysm was secured by surgical clipping and the patient recovered completely with no evidence of aneurysm formation at first follow-up imaging.

Aneurysms that originate from arteries that anastomose with the PICA are rarely described. Some of these arteries are the bulbar artery[1], leptomeningeal collateralization[2] and LSA[3-5]. The case reported here is remarkable, because the LSA was the most important anatomized vessel to take over the PICA territory supply, leading to an increased flow condition from its origin to the level of anastomosis with the PICA, evolving into an aneurysm formation and rupture as a result. The contribution to collateral supply from the SCA and AICA was less important.

The LSA has its origin from the PICA or the intradural vertebral artery, playing the role of an intersegmental anastomotic vessel, which runs posterior to the dentate ligament and anterior to the

posterior spinal roots, connecting to the posterolateral spinal artery at mid cervical level[6,7]. Many variations of the LSA with the surrounding arteries exist, which makes the interpretation of the DSA of the craniocervical region challenging[7]. In our patient we clearly identified the proximal PICA segments with normal diameter but without patent flow, indicating an old occlusion. The distal part of the telovelotonsillary segment and its vermian as well as hemispheric branches were instead filled through the LSA. Taken the preoperative imaging and intraoperative findings into account, one may hypothesize, that the flow in the proximal PICA has decreased due to stenosis or dissection with subsequent occlusion, causing cerebellar embolic infarcts and allowing for activation of the anastomotic network. The increased flow and thus the hemodynamic stress in the LSA induced the development of a small aneurysm which ruptured. Delayed occlusion of the PICA with activation of the LSA as collateral network has been reported only once before, indicating the rarity of this pathology[4]. Similar aneurysms on the LSA were also described, although not with evidence for newly developed occlusion of the PICA. The study of Chonan *et al.*, however, reported an aneurysm on the leptomeningeal collateral circulation connected to the distal PICA, with occlusion of both vertebral arteries. It is unclear whether this collateralization has developed over time, or whether it was a developmental variant[2].

Ruptured aneurysms of the LSA are rare and treatment is challenging due to the anatomical variations of its course. Occlusion of a neighboring artery (e.g. the PICA) can activate the anastomotic network of the LSA rapidly with formation of new anastomoses and consequently the risk for aneurysm formation. Careful follow-up in patients with an activated anastomotic network is warranted because development of new anastomoses can occur. In our case we will remain to follow up closely.

Funding sources

This research did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

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FIGURE LEGENDS

Figure 1: Native brain computed tomography (CT) showing subarachnoid hemorrhage in the left cerebellomedullary cistern (A) and small infarct in the territory of the posterior inferior cerebellar artery (B)

Figure 2: Lateral (A) and anteroposterior (B) digital subtraction angiography of the left vertebral artery. Single arrows show the lateral spinal artery with its aneurysm and double arrows the distal (telovelotonsillary) posterior inferior cerebellar artery

Figure 3 A: Intraoperative photograph with arterio-arterial connection between lateral spinal artery and telovelotonsillary segment of the posterior inferior cerebellar artery

Figure 3 BC: Intraoperative photograph (B) and indocyanine green videoangiography (C) before clipping

Figure 4: Indocyanine green videoangiography with retrograde flow in the telovelotonsillary segment of the posterior inferior cerebellar artery

Figure 5: Lateral (A) and anteroposterior (B) digital subtraction angiography of the left vertebral artery six months after aneurysm clipping with a new anastomotic connection between the lateral spinal artery and posterior inferior cerebellar artery

Figure 1

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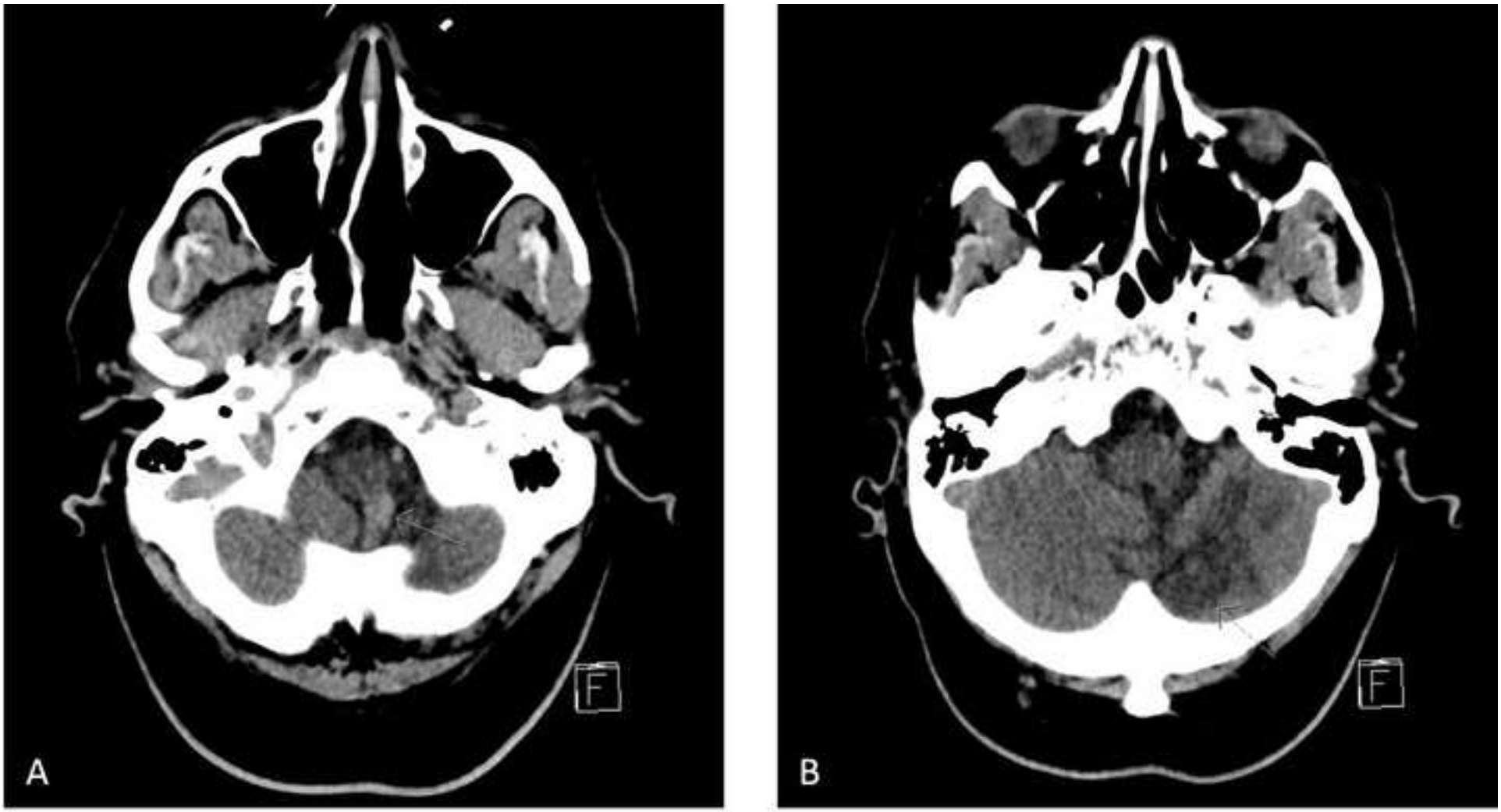

Figure 2

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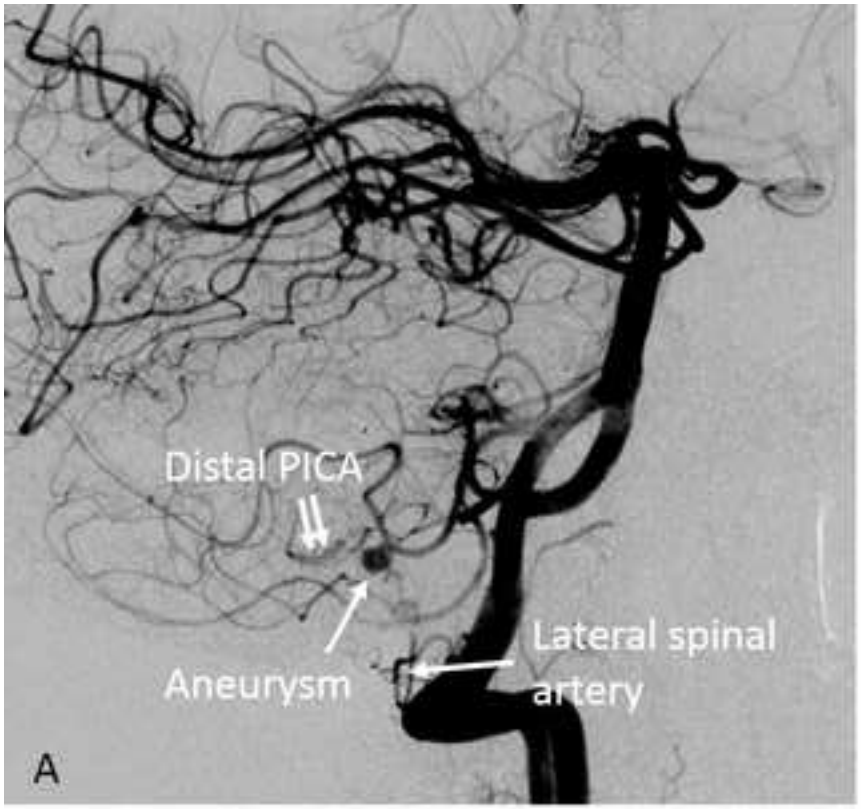

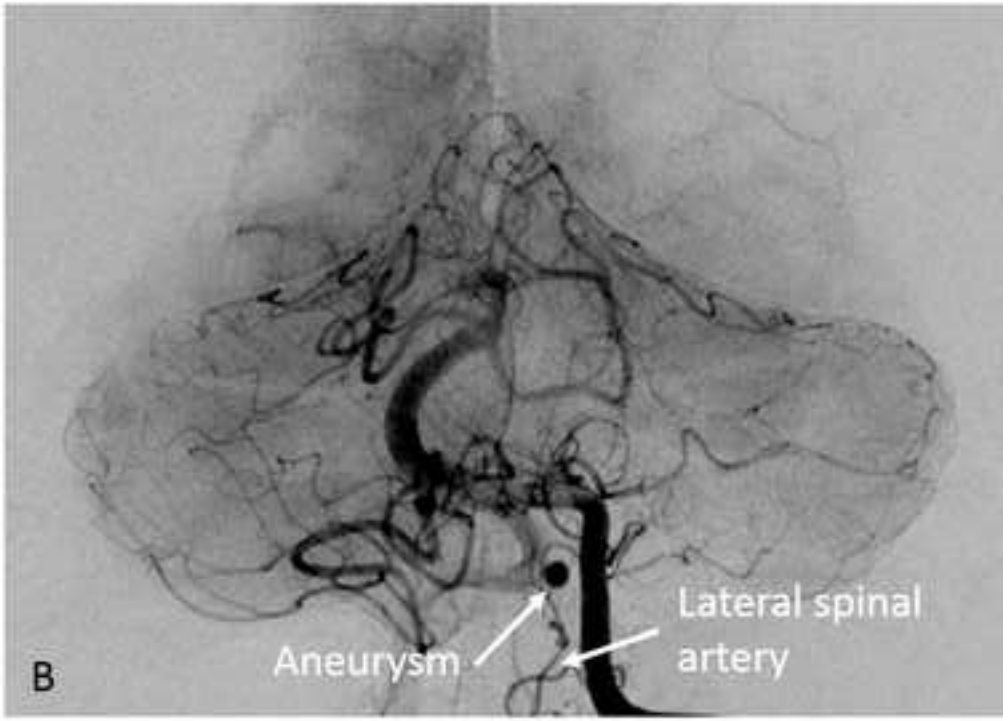

Figure 3a

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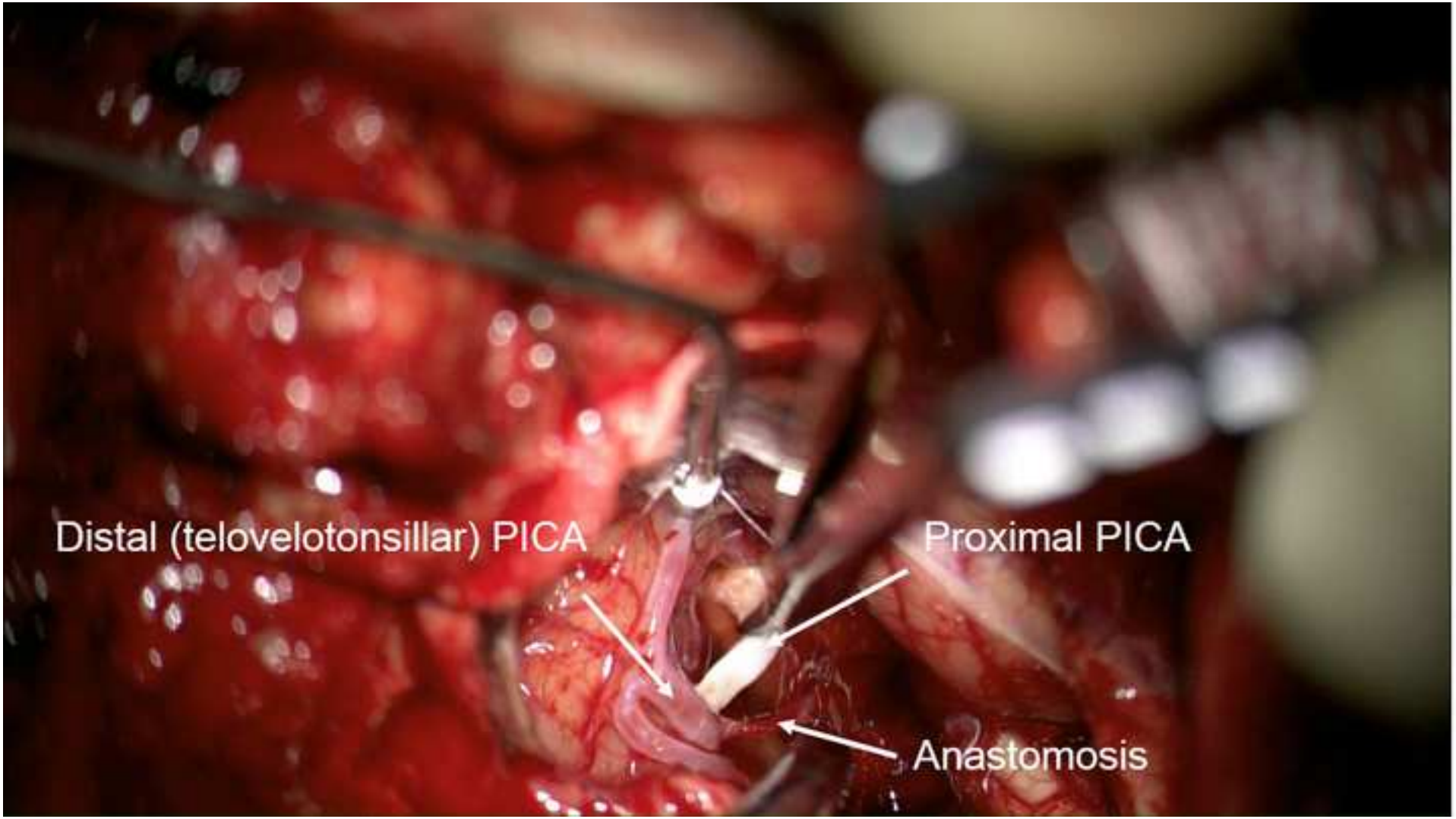

Figure 3bc
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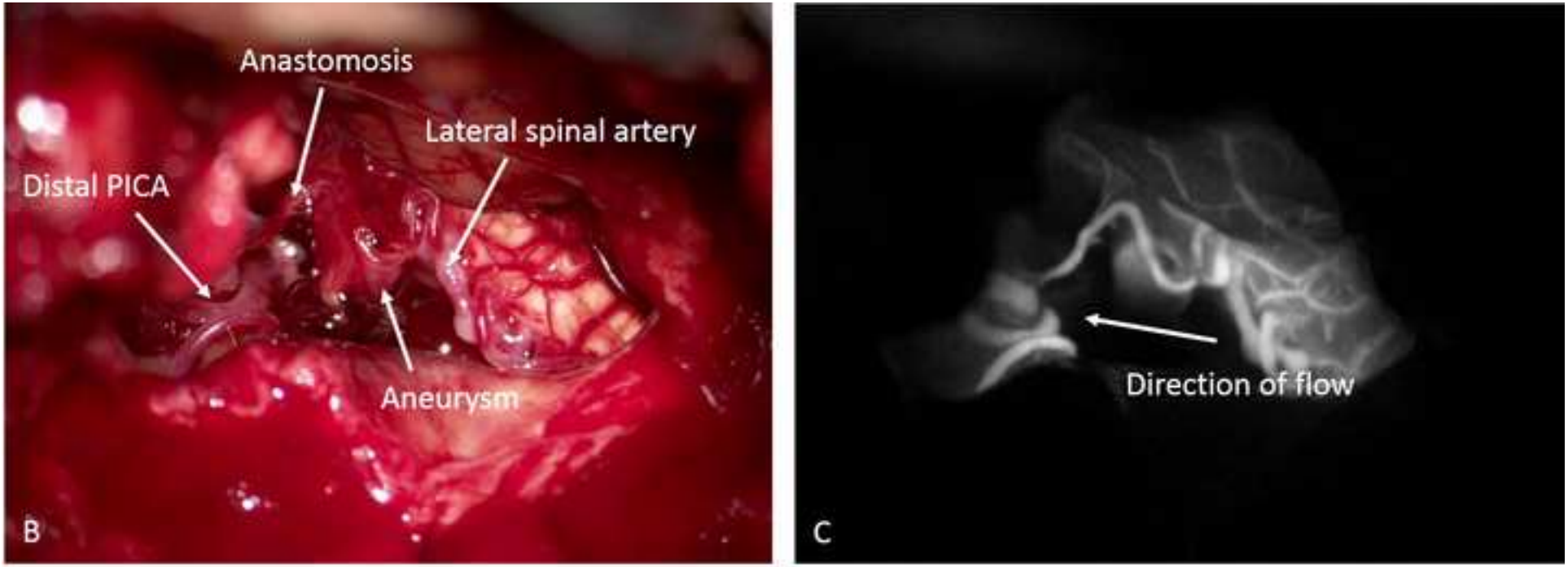

Figure 4

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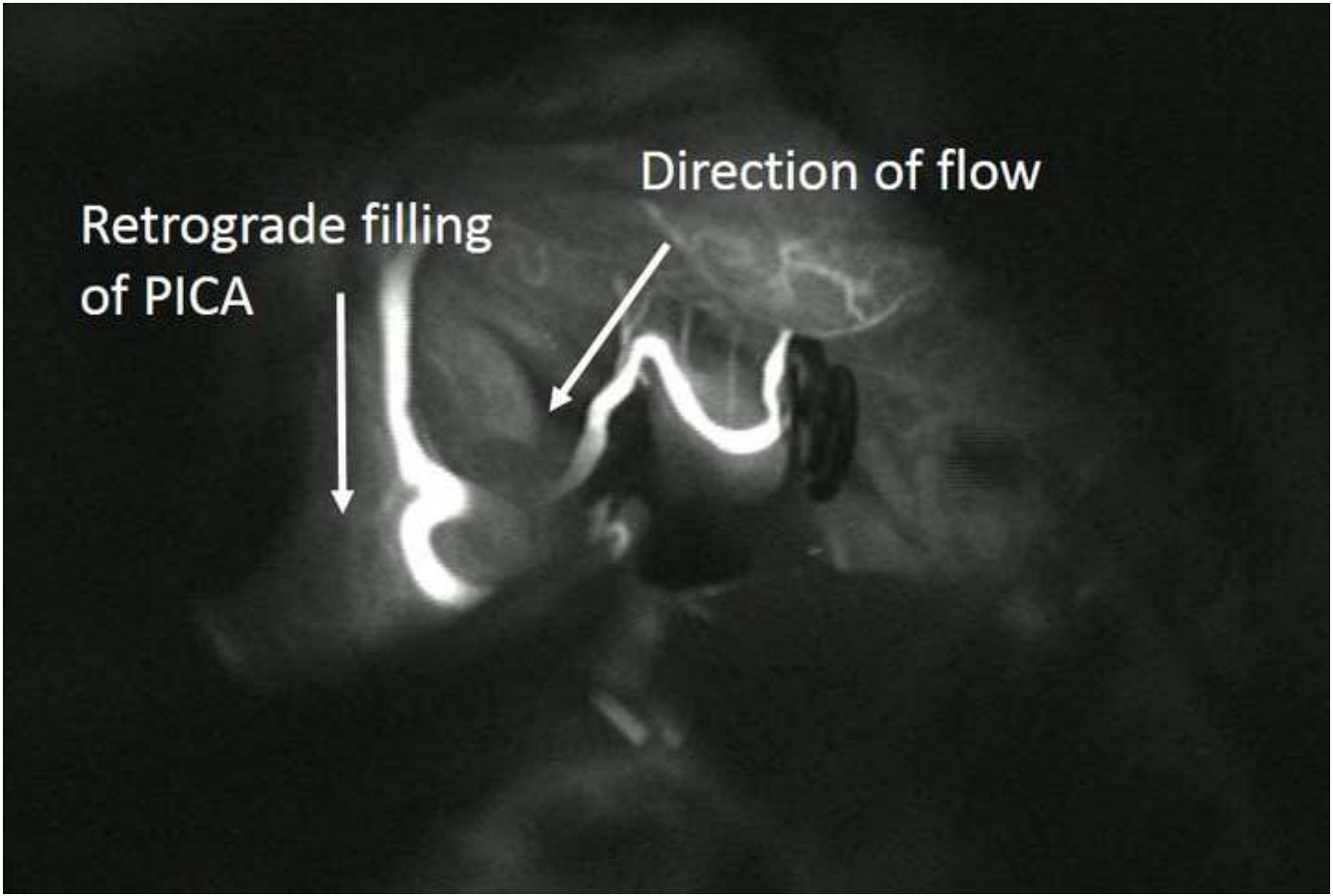

Figure 5

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